Endovascular repair of subclavian artery complex pseudoaneurysm and arteriovenous fistula with coils and Onyx

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Treatment of subclavian artery pseudoaneurysm and arteriovenous fistula (AVF) can be challenging and carries a high risk of complications. Rare cases described in the literature have been managed with major surgery or endovascular treatment with stent graft placement. We present a patient with a large lobulated pseudoaneurysm of the subclavian artery and associated high-flow AVF near the left vertebral artery origin. Surgery was considered too risky and stent graft placement would have required occlusion of the vertebral artery. Successful endovascular repair was achieved with balloon-assisted selective embolization of the fistula with a combination of coils and Onyx. (J Vasc Surg 2009;50:420-3.)

CASE REPORT

A 37-year-old woman was referred for magnetic resonance (MR) imaging/MR angiography (MRA) evaluation of her brain after new onset seizures. The patient had a history of brain infarcts dating back 19 years to a complicated childbirth. Medical records were not available for review, but the patient and her family recalled multiple central lines placed during her prolonged intensive care unit stay. On referral, the patient reported shortness of breath, progression of chronic headaches, blurry vision, increasing left-sided neck pain, and parasthesia in the bilateral upper extremities.

MR imaging of the brain demonstrated encephalomalacia as a result of chronic infarcts within the bilateral paramedian parietal and occipital lobes and the superior cerebellum (Fig 1, *A, B*). No acute findings of ischemia were noted. An MRA scan of the brain vessels demonstrated normal arteries and abnormal high signal within the left transverse and sigmoid sinuses, suggestive of arterialized blood flow.

An arteriovenous fistula (AVF) or arteriovenous malformation was suspected. An MRA scan of the neck was obtained and demonstrated a large pseudoaneurysm at the level of the left subclavian artery (SA), with retrograde high flow within the left internal jugular vein (IJV) and abnormal flow signal within the epidural space of the spinal canal at the cervical level (Fig 1, *C*). Diagnostic catheter angiography confirmed a large lobulated aneurysm of the SA and a high-flow fistula originating just distal to the left vertebral artery origin. Abrupt occlusion of the left brachiocephalic vein was seen, with no communication with the superior vena cava. Instead, venous outflow was antegrade into the enlarged left superior intercostal vein and in the hemiazygous vein, and also retrograde in the left IJV, across the transverse sinus intracranially into the right IJV as well as in the epidural venous plexus through segmental veins (Fig 2). Computer tomography (CT) brain perfusion imaging demonstrated perfusion abnormality suggestive of venous hypertension, which explained the patient's venous infarcts, headaches, blurry vision, and seizures. The patient also had edema of her left arm and decreased distal pulse.

Vascular surgeons were consulted on the patient's status. Open surgical repair would require thoracotomy or clavicular resection and carried a high risk of stroke because of the proximity of the fistula and pseudoaneurysm to the vertebral artery and the potential of injury to the brachial plexus (Fig 1, *D*). Endovascular treatment could be achieved with stent graft placement, which would occlude the left vertebral artery along with the fistula.

A decision was made to attempt selective occlusion of the fistula with coils to preserve the vertebral artery. The patient was placed under general anesthesia. A 7F Shuttle guide catheter (Cook Inc, Bloomington, Ind) was placed through the right common femoral artery into the proximal left SA. An angiogram was obtained (Fig 3, *A, B*). A Rebar 21 microcatheter (ev3 Neurovascular, Irvine, Calif) was placed coaxially into the pseudoaneurysm. An initial attempt was made to place a 14-mm × 20-cm Interlock coil (Boston Scientific, Natick, Mass). As the first coil loops were pushed out through the microcatheter, it became clear that the coil would not stay within the proximal aspect of the aneurysm and would be displaced distally. A 7 × 7-mm hyperform balloon (ev3 Neurovascular) was placed into the fistula origin. The balloon was inflated within the fistulous connection. With the balloon inflated, the rest of the coil loops were pushed into the proximal aspect of the aneurysm. This was followed by an additional two interlock coils and three hydrocoils (Microvention, Aliso Viejo, Calif) placement. To stabilize the coils, 1 cc of a non adhesive liquid embolic agent (Onyx 34; ev3 Neurovascular) was injected and distributed between the coil loops and into the proximal aspect of the fistula as the balloon was slowly retrieved and deflated (Fig 3, *C*). A final angiogram demonstrated occlusion of the fistula with preservation of the vertebral and subclavian arteries (Fig 3, *D*).

The patient was extubated without change in neurologic status and discharged the next day. Follow-up CT demonstrated normalization of brain perfusion. The patient was seen in the clinic 2 months after the procedure. Her headaches and neck pain were completely resolved and her vision improved. Left arm distal pulse remained slightly weaker. No shortness of breath.

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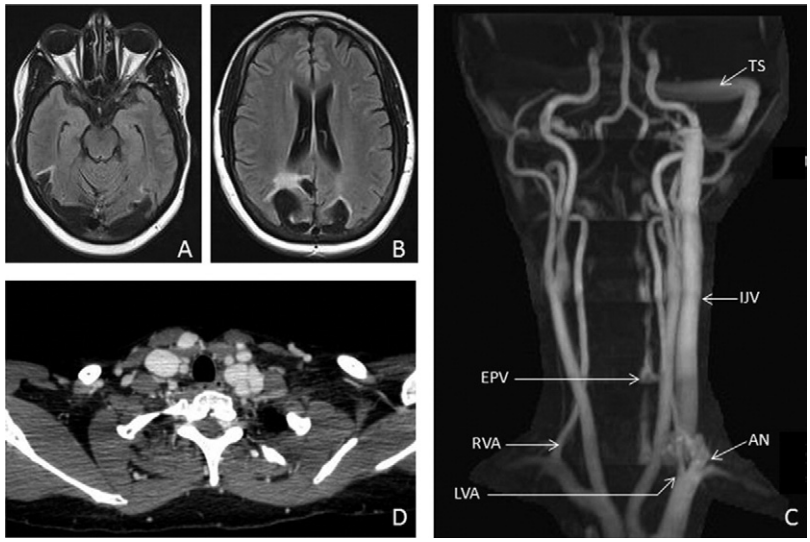

Fig 1. A, B, Magnetic resonance imaging (MRI) of the brain. Axial fluid attenuated inversion recovery (FLAIR) images demonstrate encephalomalacia resulting from old infarcts within bilateral paramedian parietal and occipital lobes and superior cerebellum. **C,** Magnetic resonance angiography (MRA). Time-of-flight technique demonstrates abnormal high signal within the left internal jugular vein (IJV), sigmoid and transverse sinuses (TS), and cervical epidural plexus venous (EPV) as a result of retrograde arterialized flow. A large aneurysm (AN) is present at the level of the left vertebral artery origin (LVA). Right vertebral artery (RVA) is normal. **D,** Computed tomography (CT) with contrast at the level of the clavicles demonstrates a large lobulated aneurysm anterior to the subclavian artery and prominent bilateral veins.

Fig 2. Three-dimension (3D) angiogram of the left subclavian artery (SA), oblique projection, demonstrates high-flow arteriovenous fistula just distal to the left vertebral artery (VA) origin, combined with a large lobulated pseudoaneurysm. Rapid filling of the remnant of the left brachiocephalic vein occluded distally and draining into hemiazygous vein (HAV) through hypertrophied superior costal vein, epidural veins (EPIV), and retrograde flow into internal jugular vein (IJ) and subclavian vein.

DISCUSSION

Iatrogenic SA-to-brachiocephalic vein fistula is a rare complication of subclavian venipuncture, with an incidence estimated to be <0.58%, most often resulting from central line placement.[1,2] If untreated over the long term, these cases are complicated by high-flow states that cause high-output congestive heart failure. In our patient, the left-sided subclavian AVF was further complicated by a large pseudoaneurysm formation and brachiocephalic vein occlusion with retrograde flow into intracranial venous sinuses resulting in intracranial venous hypertension. Another rare complication that has been reported is radiculopathy from engorgement of veins that compress the cervical nerve roots traversing the neck.[3]

Several treatment options for pseudoaneurysms and AVFs have been described in the literature. Endovascular therapy with endoluminal placement of a stent graft has been the first line of treatment.[4-6] Du Toit et al[4] reported significant stenosis or occlusion in 32% of patients followed for a mean of 49 months after endovascular repair. None of these patients experienced limb- or life-threatening complications over the follow-up period. Although these results are promising, the long-term durability of stent grafts remains a major concern. Most cases of AVF treated with endovascular repair have been the result of trauma to the SA, and few have reported complicating factors such as a pseudoaneurysm. Fausto et al[5] reported one such case with successful occlusion of the pseudoaneurysm and AVF with a fully expandable stent graft in a hemodynamically stable patient.

Surgical repair of the SA, in particular the intrathoracic segment, requires a supraclavicular incision with clavicular resection or a thoracotomy. Kalakuntla et al[7] reported a postoperative complication rate of 24%. Mortality up to 30% has been reported.

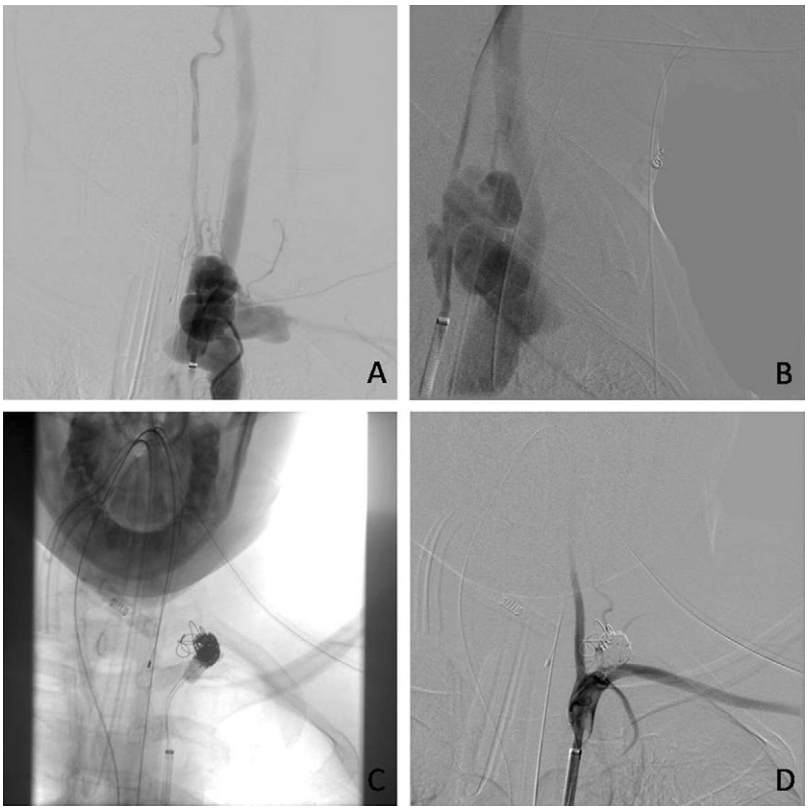

Fig 3. A, B, Left subclavian artery angiogram, anterior-posterior and lateral projections. **C,** Balloon-assisted embolization with coils and Onyx. **D,** Postembolization angiogram demonstrates occlusion of the fistula with preservation of left vertebral and subclavian arteries.

Onyx (ev3 Neurovascular) is a liquid embolic agent approved by the Food & Drug Administration (FDA) in July 2005 for the pre-surgical embolization of brain arteriovenous malformations (AVMs), and has become the preferred embolic agent for treatment of intracranial AVMs and dural arteriovenous malformations. Onyx is comprised of ethylene-vinyl copolymer (EVOH) dissolved in dimethyl sulfoxide (DMSO) with added micronized tantalum powder for easy visualization. Onyx is a biocompatible liquid polymer that precipitates and solidifies upon contact with blood, forming a soft, spongy embolus that can be pushed and directed through the vasculature.

Search of the literature found few cases of its use in the treatment of AVF in other areas of the body. Peynircioğlu et al[8] reported occlusion of a complex femoral high-flow AVF with Onyx in a patient after incomplete treatment with arterial stent graft. Successful treatment of a renal AVF with Onyx alone has been reported.[9] To our knowledge, no reported cases have combined detachable coils with Onyx in the treatment of a high-flow subclavian AVF complicated by a pseudoaneurysm.

We believe the complex lesion in our patient was the result of an old injury from central line placement 19 years before the current presentation. The case was even more unusual because of the associated occlusion of the left brachiocephalic vein and compromised antegrade outflow.

Selective endovascular occlusion of the fistula with preservation of the flow within vertebral and subclavian arteries was an optimal treatment in this case. The patient tolerated the procedure well and her clinical symptoms improved over a short period of time.

CONCLUSION

Although surgery and endovascular approach with stent graft placement remains the most commonly used treatment of SA fistula and pseudoaneurysm, selective endovascular occlusion may be warranted in certain cases and could be achieved with coils and liquid embolic devices, as described in our case.

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